A STUDY ON CLINICAL PROFILE, MANAGEMENT AND OUTCOME OF SACRO COCCYGEAL TERATOMA IN CHILDREN

Dissertation Submitted to

THE TAMIL NADU DR.M.G.R MEDICAL UNIVERSITY

In partial fulfillment of the requirements

for the award of the degree of

M.Ch. Branch V

PEDIATRIC SURGERY

2011 – 2014

THE TAMIL NADU DR.M.G.R.MEDICAL

UNIVERSITY, CHENNAI.

AUGUST – 2014.

CERTIFICATE

This is to certify that the dissertation entitled **"A study on the profile, management and outcome of Sacrococcygeal Teratoma in children"** is a bonafide work done by Dr. K.Sriram, under my guidance and supervision during the period between 2011-2014 towards the partial fulfillment of requirement for the award of M.Ch Branch V (Pediatric Surgery) degree examination held in August 2014 by The Tamil Nadu Dr .M.G.R. Medical University, Chennai.

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DECLARATION

I solemnly declare that the dissertation entitled **"A study on the clinical profile, management and outcome of Sacrococcygeal Teratoma in children"** is the original work done by me at the Institute of Child Health and Hospital for Children, Egmore, during the M.Ch. course (2011-2014), under the guidance and supervision of Prof. S.V. Senthilnathan M.S., M.Ch. Professor and Head of the Department of Pediatric Surgery. This dissertation is submitted to The Tamil Nadu Dr.M.G.R. Medical University, Chennai towards the partial fulfillment of requirement for the award of M.Ch. (BRANCH – V) in PAEDIATRIC SURGERY**.**

Place: Chennai. .

Date: **Dr.K.SRIRAM**

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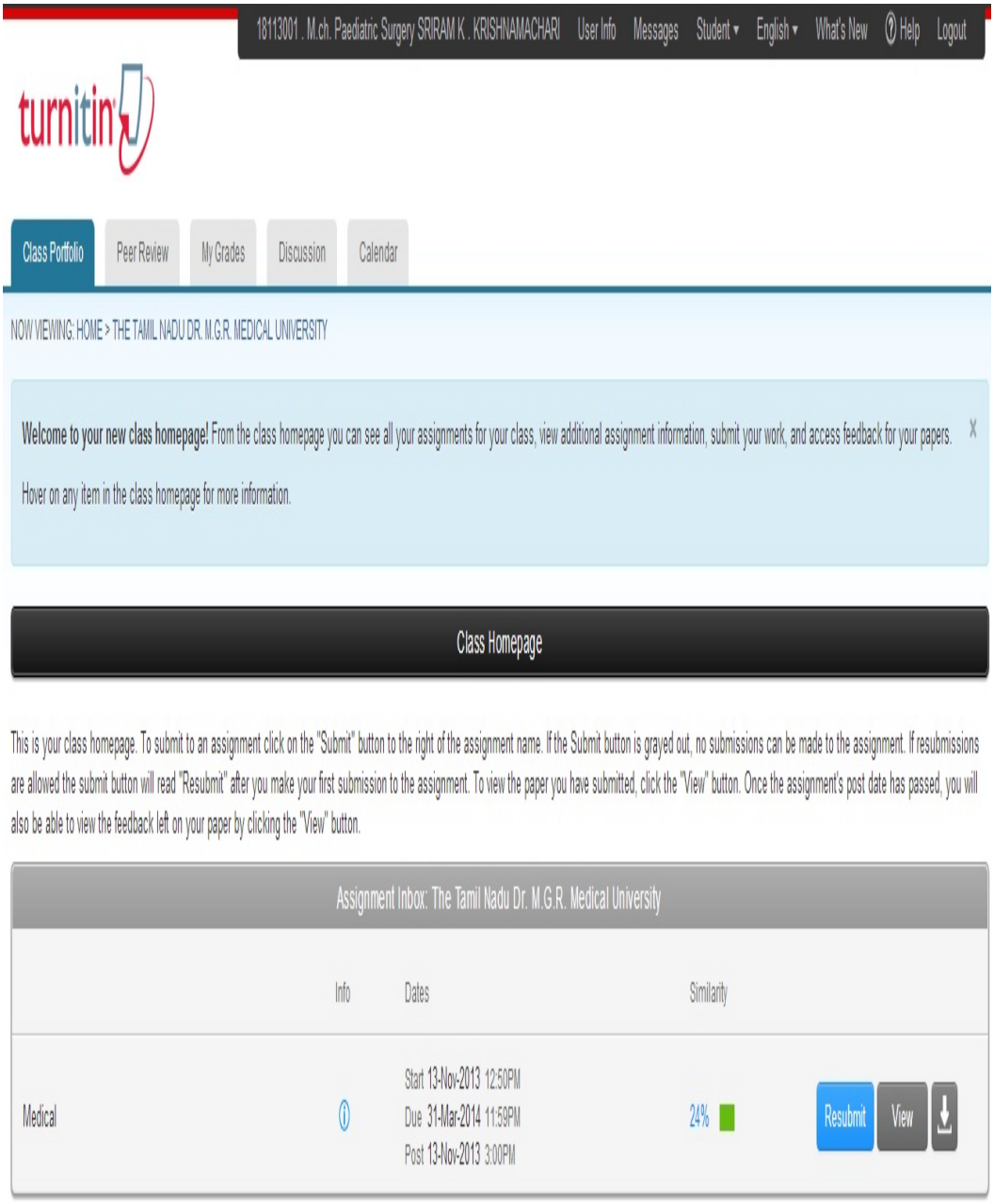
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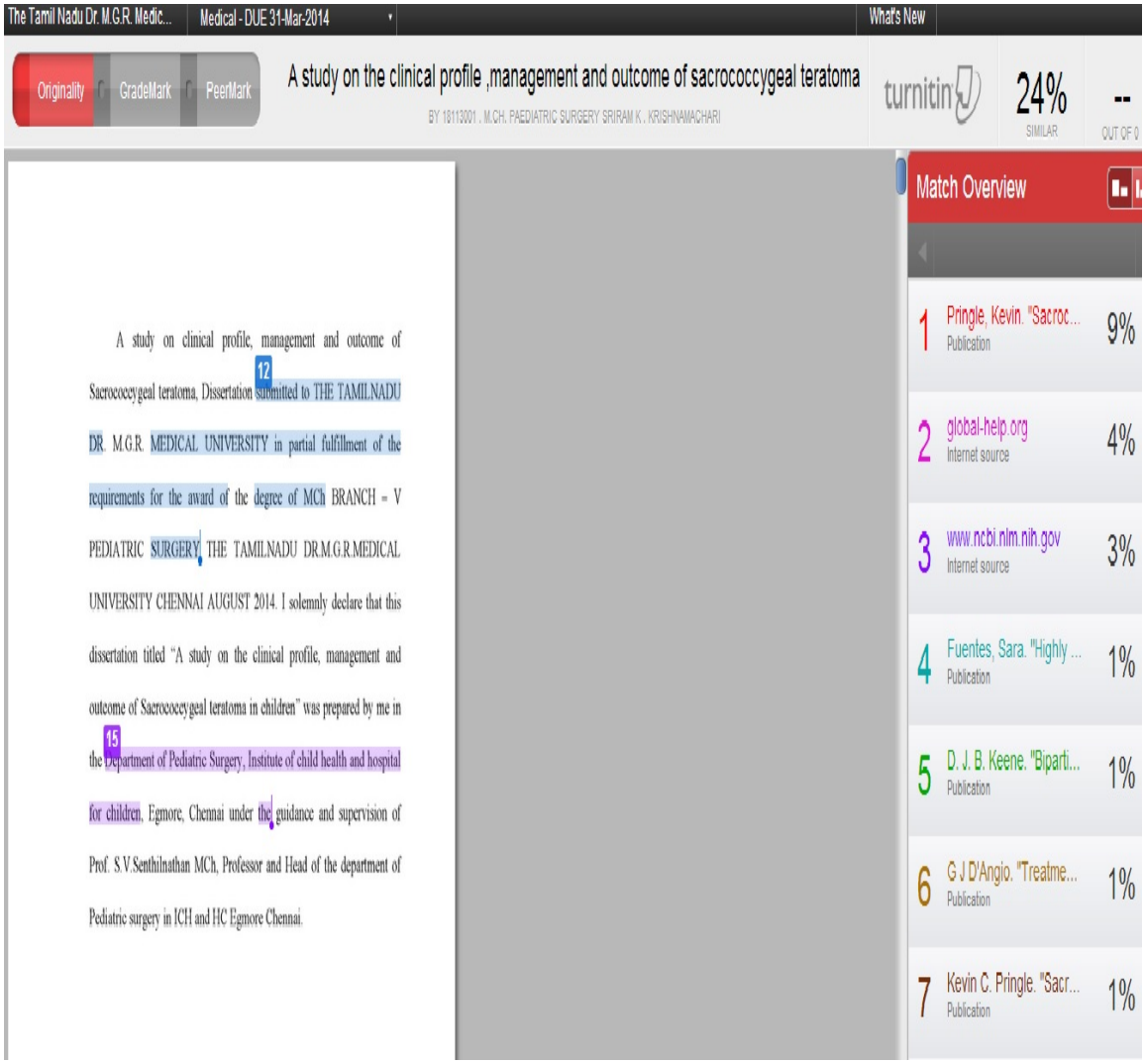
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To
Dr.K.Sriram
II year, Mch (Paeadiatric Surgery)PG,
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Dear Dr.Sriram

The Institutional Ethics committee of Madras Medical College, reviewed and discussed your application for approval of the proposal entitled "Clinical profile, management & outcome of sacrococcygeal teratoma" No.07042013.

The following members of Ethics Committee were present in the meeting held on 17.04.2013 conducted at Madras Medical College, Chennai -3.

1. Dr.G.SivaKumar, MS FICS FAIS --- Chairperson
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We approve the proposal conducted in its presented form.

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The Institutional Ethics Committee expects to be informed about the progress of the study, and SAE occurring in the course of the study, any changes in the protocol and patients information / informed consent and asks to be provided a copy of the final report.

18/4/13

Member Secretary, Ethics Committee

INDEX

Abstract

Aim:

The primary aim is to analyze the clinical profile including epidemiology, clinical presentation, diagnosis (ante natal and post natal),operative findings, and outcome

Methods:

15 Children with Sacro Coccygeal Teratoma were evaluated clinically, radiologically, and with tumour markers. Subsequently they were subjected to management protocol which included surgery, chemo therapy, and radio therapy. They were followed for 2 years

Observation and results

In our study females were more affected, surgery alone was inadequate for successful management of malignant SCT. We had 5 immature teratoma. Altman type 1 type2 were more common, 46% presented at birth, benign SCT had favorable prognosis, 13% had a pre natal diagnosis, 67% were term, renal anomalies were the most common associated anomaly.

Conclusion

SCT are a relatively uncommon tumour. Those presenting within a few days of birth carry a better prognosis. Mass is the most common presenting symptom. Early diagnosis, prompt surgical intervention gives good result and long term survival. The optimal therapeutic program for children with malignant SCT is still evolving.

KEY WORDS

Sacro Coccygeal Teratoma, Altman type, tumour marker

INTRODUCTION

Sacrococcygeal Teratoma is a neoplasm arising from the caudal end of spine, usually protruding from the inferior end of the spinal column and displacing the anus forwards.

Sacro Coccygeal Teratoma is defined as a tumor emerging from the proliferation of pluri potent cells. They contain tissues from one or more germ cell layers.

Though rare it is the most common tumor in the new born period with incidence of 1:35000-45000 live births. Male female ratio is 1: 3-4. They are composed of 2 or 3 germ cell layers, having multiple tissue types. They present in varying sizes and shapes.

Sacro Coccygeal Teratoma is tumor of new born which carries good prognosis if diagnosed early and adequate surgical treatment is prompt.

AIMS AND OBJECTIVES

The primary aim is to analyze the clinical profile including epidemiology, clinical presentation, diagnosis (ante natal and post natal), operative findings, and outcome.

REVIEW OF LITERATURE

HISTORY

Historically, teratomas were attributed to demons, sexual misconduct and abnormal fertilization.[1]

Virchow in 1869 applied the term teratos (“of the monster “) and onkoma (“swelling “).

EMBRYOLOGY

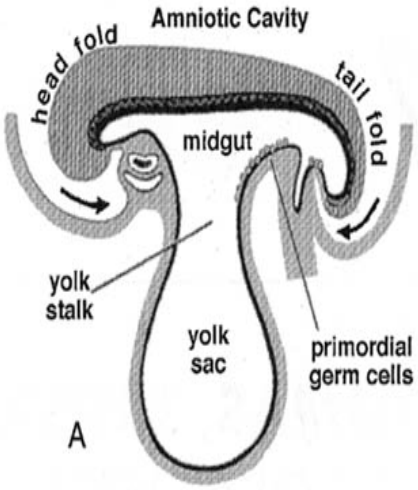
Teratoma is considered to have many tissues, foreign to the organ/site from which they are said to arise.[2]

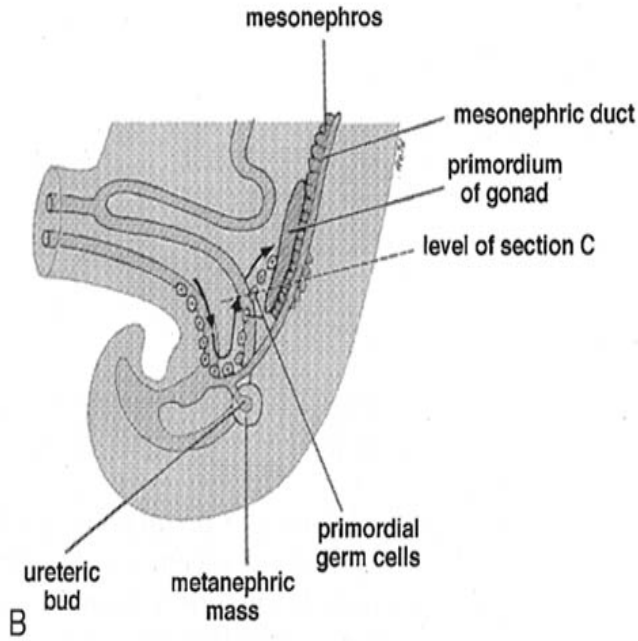
Earlier teratomas were thought to have three embryonic layers (Ectoderm, endoderm, and mesoderm).

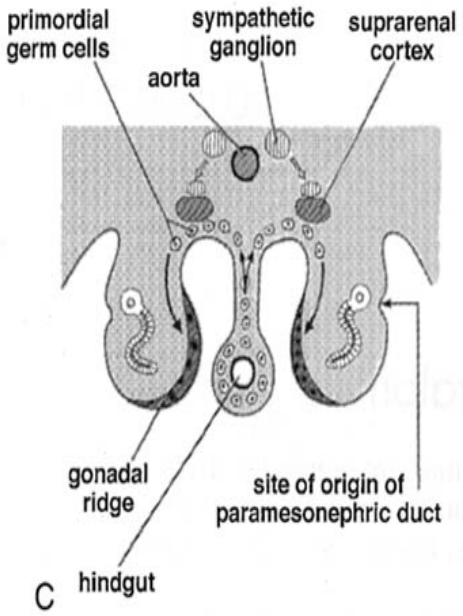
But in the recent past monodermal types have been included .[2,3]

Teratomas were perceived to arise from totipotent primordial germ cells.[3,4] They develop among the endodermal cells of the yolk sac near the origin of the allantois and migrate to the gonadal ridges during 4^{th} and 5^{th} weeks of pregnancy. [5]

EMBRYOLOGY







Some of these miss the target and give rise to teratoma anywhere, commonly in the midline.

Another proposed theory is that they arise from remnants of primitive streak/ node - Hensen node. [5-7]

During the third week of gestation, midline cells at the caudal end of embryo divide rapidly and by the processes called gastrulation give rise to all the three germ cell layers of the embryo.

By the completion of the third week, primitive streak shortens and disappears. This is said to explain the more common occurrence of teratoma in the sacro coccygeal region.

Teratomas have diversity of tissues and varying degree of organization of these tissues.

They may contain skin elements, neural tissue, teeth, fat, cartilage, and intestinal mucosa often with normal ganglion cells.

Tissue architecture is variable.

Spectrum of cellular differentiation exists.

Most benign teratoma are composed of mature cells, 20-25% also contain immature elements. (Neuro epithelium)

PATHOLOGY

Teratoma is the result of continued multiplication of totipotent cells from Hensen node which fails to involute at the end of embryonic life.

DEFINITION:

WILLIS

"A teratoma is a true tumor or neoplasm composed of multiple tissues of kinds foreign to the part in which it arises."

Most common site is the Sacro coccygeal region.

ROBBINS

'A tumor composed of cells representing more than one germ cell layer in which any one tumor cells can vary from totally benign (mature) to cells that appear frankly malignant. (Immature)

Risk of malignancy depends on

1. Age of diagnosis of the disease.
2. The site of occurrence,
3. Extent of disease.

Age greater than 2 years has high risk of malignancy.

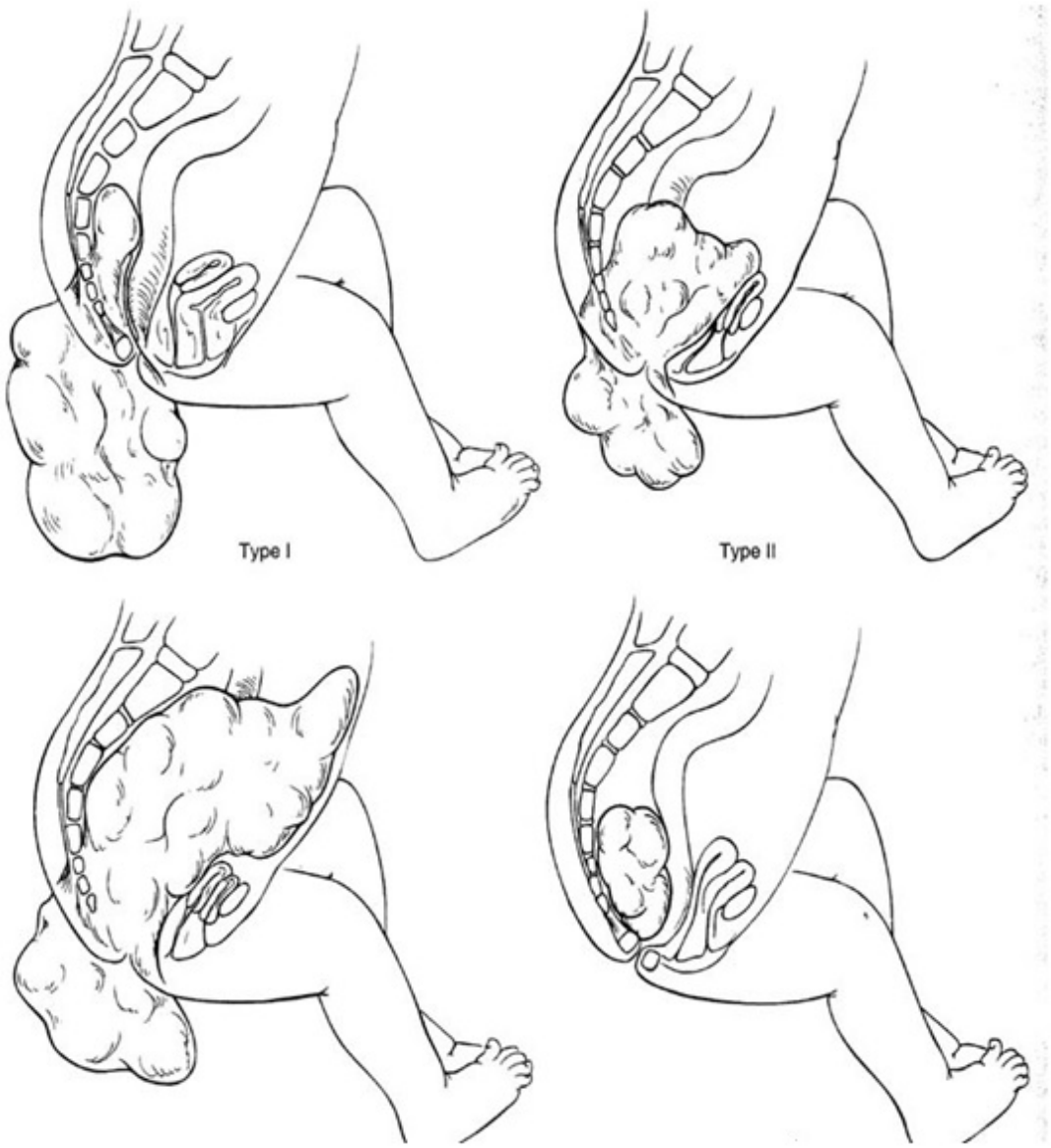
Altman et al classified Sacro coccygeal teratoma into 4 types.

1. Occur almost exclusively exterior with a minimal pelvic/presacral component.
2. Apart from the External presentation they have significant pelvic component.

3. They have intra pelvic and intra abdominal component greater than the external component.

IV They are almost exclusively Pre sacral.

ALTMAN CLASSIFICATION



Type III **Type IV**

INCIDENCE OF MALIGNANCY

Altman Type	Incidence (%)	Malignancy (%)
I	47	Rare
II	35	6
III	8	20
IV	10	8

ASSOCIATED ANOMALIES;

Sacro coccygeal Teratomas are usually isolated lesions.

They form a part of Currarino triad .The triad includes

1. Ano rectal malformation
2. Sacral anomaly
3. Pre Sacral masses. (Usually a teratoma or an Anterior Meningocele.)

Familial pre disposition is noted in 57% of cases and has an autosomal dominance.

The most common Ano rectal malformation is Anal or Ano rectal stenosis.

Other associated anomalies are

Hypospadias.

Vesico ureteric reflux.

Vaginal / uterine duplication.

Congenital dislocation of hip (7%)

Vertebral anomalies which leads to late orthopedic sequalae.

Central nervous system associated anomalies are

Anencephaly

Trigonocephalay.

Dandy Walker Malformation.

Spina Bifida.

Myelomeningocele.

Another peculiar association is with family history of twins. (10 %)

Teratomas are just one end of the spectrum of conjoined twinning.

PRESENTATION:

Post natal

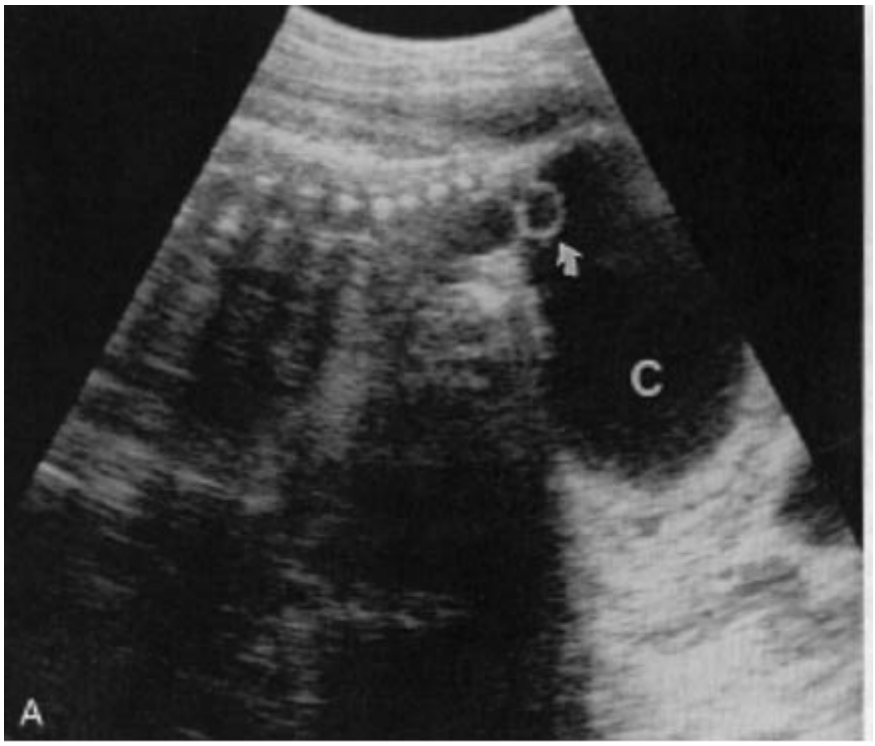
The most common presentation is a large sacral mass seen at birth.

Malignant tumor is more common when the child presents at 5 – 6 months of age as a swelling in the buttock.

Ante natal

Recently most cases are detected by routine antenatal ultrasonogram

ANTE NATAL ULTRASOUND



Majority of them die before birth as mostly they are as great as or greater than the rest of the fetus. They die from heart failure as the fetus heart is not able to pump sufficient blood to both the fetus and the mass.

In most of the antenatal series

Fetal hydrops/ non immune are very common and are associated with increased incidence of fetal death. [8,9,10,11-16]

DURING LABOR

Another mode of presentation is when the tumor becomes impacted during delivery causing death of the fetus by obstructing delivery or tumor ruptures during delivery and the infant bleeds to death shortly after birth.

CLINICAL FEATURES.

The commonest mode of presentation to the pediatric surgical department is a large skin covered mass protruding from the coccygeal region, pushing the anus and vagina anteriorly. They have large veins on the surface. Large tumors may have ruptured / bled or have ulcer on the surface.

Urinary tract obstruction and constipation symptoms precede Sacro coccygeal Teratoma detection in infants.

COMMONEST PRESENTATION -MASS

Malignant tumor commonly present as a rapidly growing buttock mass. Distant metastases at presentation are rare due to early diagnosis by ante natal ultrasonogram.

Risk of malignancy is

< 10 % at birth

>75 % at one year of age.

Children and adolescent with pre sacral mass present with constipation and urinary retention.[17,18,19-21]

Diagnosis of pure intra pelvic teratoma is delayed because of non specific symptoms like constipation, urinary retention, abdominal mass, and symptoms of malignancy (failure to thrive).

Lesion with large intra pelvic component may cause urinary obstruction.

Retro rectal mass is recognized on per rectal examination as solid pre sacral mass. The Tumor is firmly attached to or arises from the anterior surface of the coccyx. The coccyx is displaced posteriorly. The sacrum is normal.

Blood supply is from the middle sacral artery.

DIFFERENTIAL DIAGNOSIS.

1. Anterior meningocele: It is cephalad to sacrum and is covered by duramater. During examination when pressure is applied on the mass it causes a bulge in the anterior fontanelle. (Positive cross fluctuation). On per rectal examination a cystic mass can be felt and the anterior sacral defect can also be palpated.

2. Sacro coccygeal chordoma.

3. Lymphangioma

4. Lipoma

5. Tail like remnants.

6. Meconium pseudo cysts.

DIAGNOSIS:

Presents as a visible mass at birth.

Many present as asymptomatic mass but some present with high output cardiac failure, Disseminated intravascular coagulation, rupture, bleeding, lethal hyperkalemia (due to tumor necrosis) and prematurity requiring intensive care management.

Plain X Ray of pelvis with spine: AP & lateral

1. Differentiates from anterior meningocele which has a characteristic defect in the sacrum.

2. Tumor calcification.

3. Spinal defects.

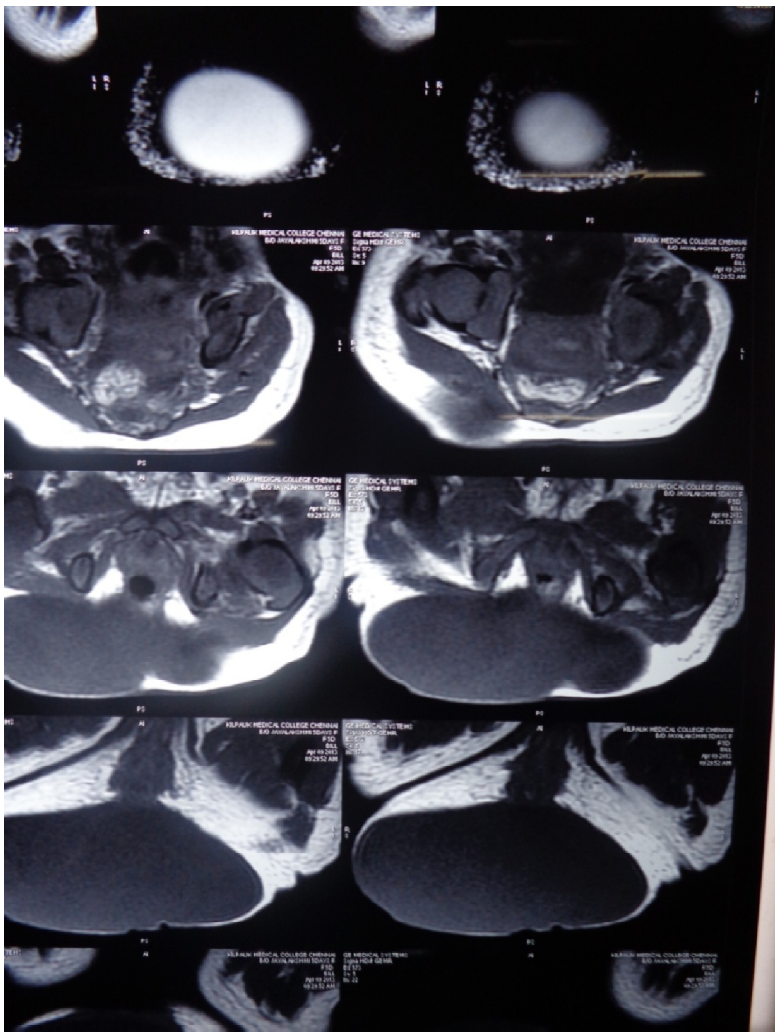
Ultra sonogram of abdomen, pelvis & spine:

Gives an assessment of the size, consistency of any pelvic or abdominal component.

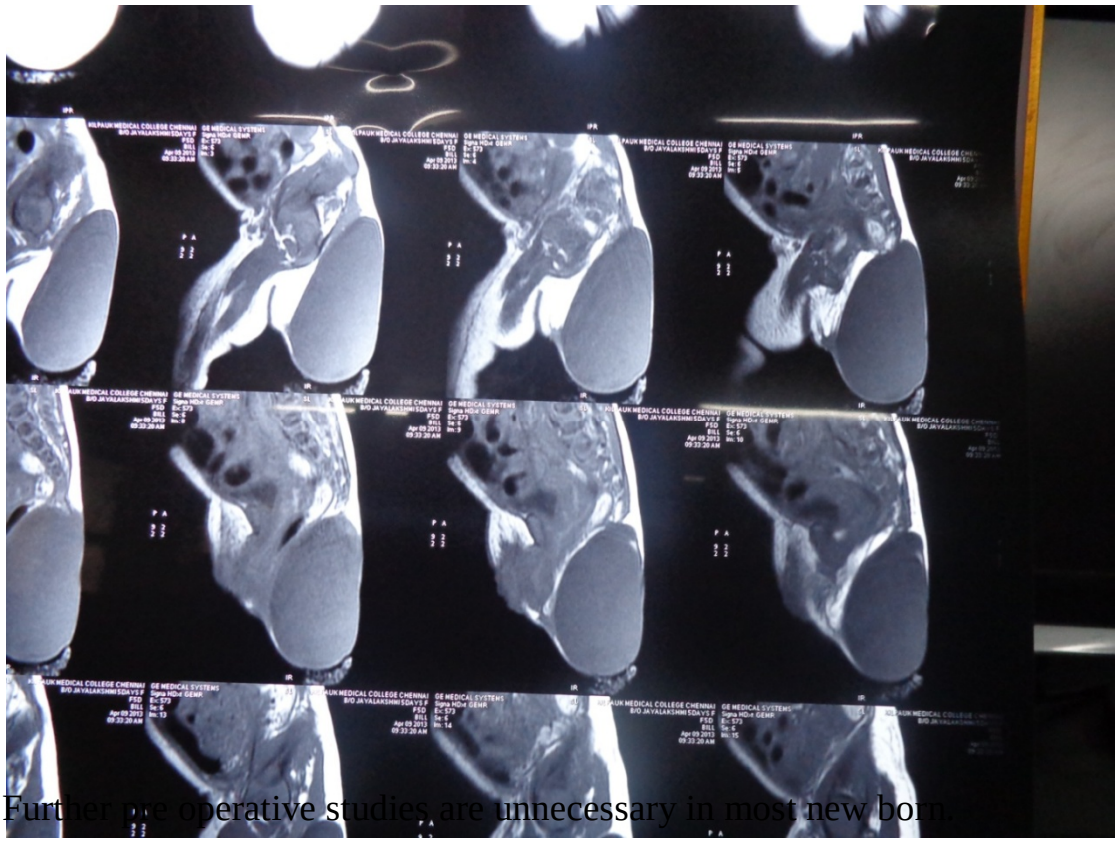
MAGNETIC RESONANCE IMAGING:

It can be used in neonates with minimal sedation but most require general anesthesia. It delineates the vascular anatomy, [22-24] Oil instilled into rectum during the scan can be used as a contrast medium.[25,26] It clearly distinguishes Sacrococcygeal teratoma from Anterior meningocele. It can also detect occasional extension of the tumor through sacral hiatus into spinal canal.[27]

MRI PELVIS: T1 IMAGES



MRI PELVIS: T2 IMAGES



Further pre operative studies are unnecessary in most new born.

PRE NATAL DIAGNOSIS

Second trimester ultra sonogram defines the site of the lesion, complex appearance, intra pelvic extension with or without urinary tract obstruction.

Presence of a large solid vascular component is associated with significant mortality both in utero and perinatal period.[28-31]

Ante natal MRI:

Defines the anatomy of the tumor, and the blood supply to the tumor in utero. There is no need for fetal sedation or paralysis

Perinatal mortality is related to pre maturity, tumor rupture with exsanguinations or both.

TABLE 38-3. *Normal infant serum α-fetoprotein (AFP) levels at various ages*

Age	Mean AFP (ng/mL)
Premature	134,000
Newborn	48,000
Newborn to 2 wk	33,000
2 wk–1 mo	9,500
2 mo	323
3 mo	88
4 mo	74
5 mo	47
6 mo	13
8 mo	8.5

NATURAL COURSE:

Pre mature delivery may occur spontaneously from polyhydramnios or may be induced urgently because of fetal distress.

Repeat ultra sonogram is a must for ante natally detected tumors.

Lower segment caesarean section is considered if tumor is greater than 5 cm or greater than fetal Bi parietal Diameter. This is to avoid dystocia during vaginal delivery and is an obstetrical night mare which is avoidable.

Management of unexpected cases with dystocia is emergency caesarian section, completion of partially delivered fetus who has been intubated and ventilated after vaginal presentation of head.

MANAGEMENT

ANTENATAL

Fetal MRI is an adjunct to pre natal evaluation to provide additional information that helps in counseling and pre operative planning [52]

Fetal MRI also helps in differential diagnosis when the lesion is entirely pre sacral.[53]

Intra uterine laser ablation can be done [54]

The improved diagnosis and almost uniform mortality rate associated with fetal hydrops have provided considerable impetus to consider fetal surgery or per cutaneaous shunting or drainage to allow vaginal delivery for selected cases of ante natally diagnosed sacro coccygeal teratoma. But the results are mixed.

Interrupting the high vascular flow to the tumors with radio frequency ablation.[55]

Per cutaneaous aspiration to facilitate delivery[31,56] to eradicate uterine irritability or prevent tumor rupture at delivery.

Pre natal decompression using cyto amniotic shunt to relieve obstructive uropathy.

It would be fair to say that the role of fetal intervention in the presence of this tumor is not well defined.

POST NATAL MANAGEMENT:

Recently multi modality treatment approach has resulted in improved survival of more than 80% of sacrococcygeal teratoma.

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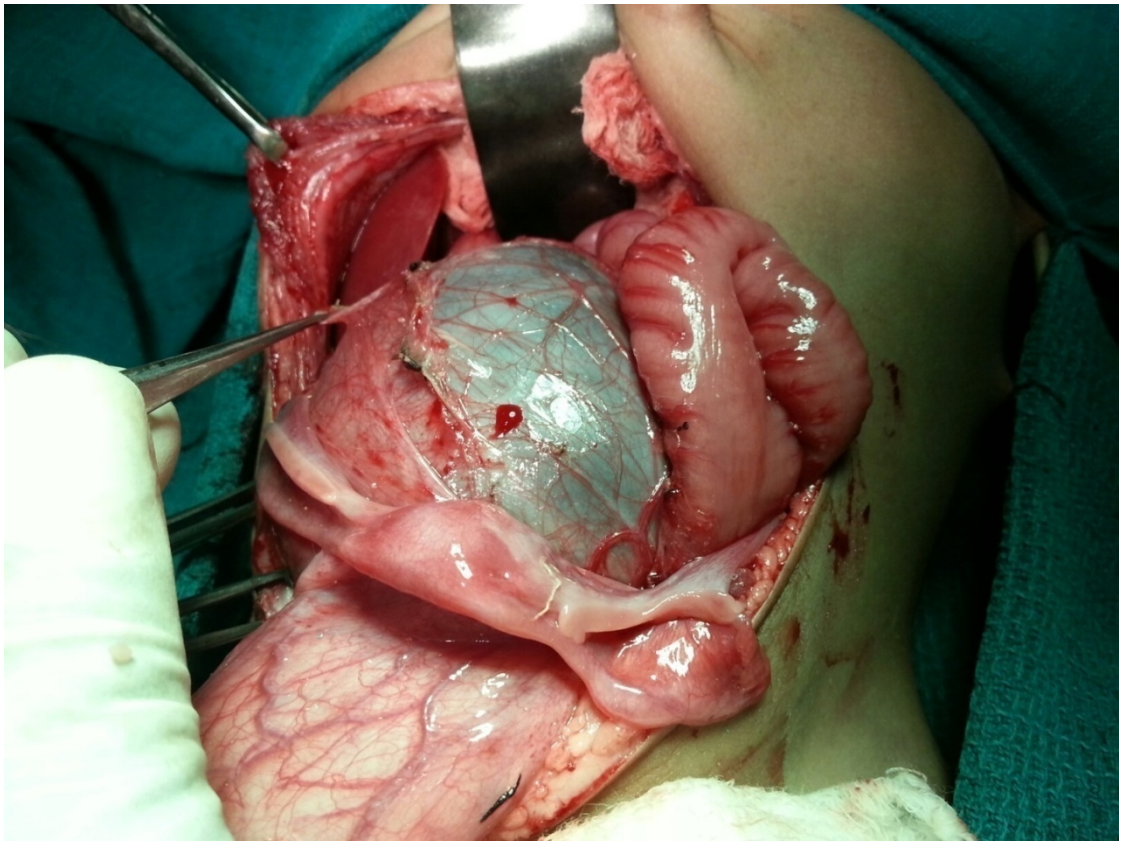
Preliminary abdominal procedure is to be considered in case of a very large tumor.

ALTMAN TYPE III – ABDOMINAL APPROACH



Laparoscopic procedure to divide the middle sacral artery has also been described.

Complete excision with coccygectomy is done as soon as the neonate is stable enough to undergo the surgery.

Serum tumor marker is done before surgery and later for comparison.

PRE OPERATIVE MANAGEMENT:

If the tumor is intact and the child stable then there is no need for an emergency resection. Surgery done within the first 24 hours of birth has a good prognosis and the rate of complication is less as the gut is usually not colonized in the first 24 hours of life. Early resection decreases the rate of infection even if the field is contaminated by stool during resection as the gut is not colonized in the first 24 hours of life. The new born is started on antibiotics immediately and the same has to continue for 24 – 48 hours post operation. Bowel preparation formally has to be done if the infant is several days old or has been fed.

Blood for alpha feta protein is to be taken as a base line in order to confirm post operatively that its level falls at the normal rate.[10,57-59] Rarely alpha feta protein might not be elevated. Adequate cross matched blood has to be reserved before surgery. Good venous access has to be obtained. Consider an arterial line if possible. If tumor ruptures, then pressure bandage may stem blood loss but carries the risk of “squeezing” immature cells into the venous drainage and lodge these cells in lungs. Metastatic disease is not a problem in these infants. Emergency surgery is indicated in these circumstances. When the mass is as large as a fetus, one should not wait for the baby to grow, but the tumor has to be resected

as the incidence of malignancy is said to increase with age more particularly if the infant is 2 month old.

SURGICAL PROCEDURE:

BLADDER CATHERIZED AND RECTUM PACKED

The child is placed in the prone position with a roll under the hips. The bladder is catheterized. The anus /rectum are packed with a betadine gauze pack. The anus is prepped out of the field.[60,61] The incision is a chevron or a vertical on the dorsum of the mass preserving as much of

the skin as possible. Excess skin can always be trimmed later. Large veins in the sub cutaneaous plane are divided between ties. The apex of the chevron should be situated over the sacrum.

POSITION ON OPERATING TABLE

If significant intra pelvic or intra abdominal component is present, begin with laparotomy /laparoscopy. Preliminary abdominal exploration is also indicated when

1. There is rupture or bleeding,

2. Pre mature baby in hyper dynamic state and

3. When preliminary devascularization is needed to stabilize the child.

En bloc excision including coccyx is preferable. Failure to excise the coccyx is associated with a high recurrence rate.[67-69]

OPERATIVE STEPS

The options available for the management of large sacro coccygeal teratoma are

1. Intra operative snaring of aorta [62,63]

2. Laparoscopic clipping of middle sacral artery.[64]

3. Use of ECMO and hypothermic perfusion.

4. Devascularization and staged resection [65]

5. Pre operative embolization.

6. Radio frequency ablation.

7. Autologous cord blood transfusion is a useful adjunct.[66]

POST OPERATIVE MANAGEMENT:

The child is nursed in the prone position. The urinary catheter is removed as soon as the child is stable. The child is fed as soon as it is extubated. The drain is removed in the first few days. Alpha feta protein is determined in the immediate post operative period and on discharge.

In the older patients, treatment of the malignant tumor is excision and chemotherapy and monitoring with imaging studies and serum markers.

For unresectable tumors biopsy and chemotherapy followed by excision of the primary tumor after adequate reduction in the size of the tumors has been obtained.[44]

Radiation therapy is usually reserved for local recurrence of the malignant tumor. The child with malignant tumor has be be enrolled in pediatric co operative study or treat according to their guidelines.

COMPLICATIONS OF SURGERY:

1. Nocturnal enuresis.
2. Perineal anesthesia
3. Patulous anus.
4. Neurogenic bladder.
5. Urinary and fecal incontinence.

Modern chemotherapy has produced very good improvement in survival.[41,42] Survival rates as high as 80 %has been noted but the risk of late recurrence or second cancer persists. Functional results in survivors are related to fecal and urinary continence problems. Good outcome requires meticulous dissection along tumor capsule, preservation/reconstruction of muscular structures.

FOLLOW UP/MONITOR:

Physical examination and per rectal examination and tumor markers.

Every 2 months for 3 years because most recurrence occur in 3 years of surgery. [43]

In the long term follow up early assessment of bladder, anorectal and sexual function along with cosmetic results within a structured oncology follow up program is ideal.

At monthly intervals for 3 months.

At three monthly intervals for 1 year.

At six monthly intervals for 1-3 years.

At yearly intervals for 3- 5 years.

At each visit the following procedures are performed,

1. Per rectal examination to rule out local recurrence

2. Serum alpha feta protein for distant spread.

3. Renal ultra sonogram.

Follow up continues for 5 years.

PROGNOSIS:

Good Prognostic factors

1. Fetus with tumors diagnosed in utero, the survival is greater than 90 %, if it is diagnosed by routine ultra sonogram.

2. Tumour is small.

Bad Prognostic factors

1. When polyhydramnios with a larger tumor is present.

2. When tumor is larger than fetal bi parietal diameter.

3. As tumor enlarges, fetus develops placentomegaly or hydrops which are harbinger of impending fetal death. This should lead to urgent Lower segment caesarian section especially when the maternal mirror syndrome is present. [51]

In the absence of distal metastasis at presentation and if surgery has removed the complete tumor, life expectancy is considered to be normal.

Prognosis for malignant Sacrococcygeal Teratoma is not good and guarded.

The mortality increases by 60%if the pregnancy is complicated which has been an indication of ultrasonographic evaluation.

The mortality is 100 % if hydrops or placentamegaly occurs.[29-31]

The mortality is said to be 50% if the size of the tumor is 10 cm or greater that were highly vascular or fast growing.

In the absence of severe prematurity and intra partum complication, prognosis depends on presence of malignancy and is hence related to the age of operation and completeness of excision.

Cystic lesions are benign but the presence of solid component indicates malignancy.

The risk of recurrence is increased with the findings of immature histology and incomplete resection.

Although malignant recurrence of a benign teratoma may be as high as 10 – 19%.[7,32] the original benign diagnosis may have been due to sampling error, [33] an undetected residual microscopic focus of malignant tumor [34] or secondary to incomplete coccygectomy at the initial operation. [35]

Those tumors which are resected after the new borne period have a higher risk of malignant recurrence, especially when an elevated alpha protein is present at diagnosis.

Elevated alpha feta protein signifies the presence of malignancy in the original tumor.[33,37]

Prognosis is not dependent on Altman classification [38] but rather on tumor size, physiologic consequences, histology, and associated anomalies. The prognosis of malignant tumor depends on tumor type, stage [39]location and patient age. The recurrent disease is usually local, but metastasis occurs to inguinal nodes, lungs, liver, brain and peritoneum (pseudomyxmo peritonei). [40] The prognosis of malignant tumor was dismal till the advent of platinum based chemotherapy.[34]

MATERIALS AND METHODS:

Study population: All patients who presented to our hospital with sacrococcygeal teratoma.

Number of cases: 15.

Study period: December 2011 to December 2013.

Exclusion Criteria: All sacro coccygeal tumors not proven to be teratoma e.g. Hind gut duplication cyst, Dermoid, Meningocele, Pelvic tumors – Neuro blastoma.

Screening Procedure: All cases of sacro-coccygeal tumors were examined in detail and subjected to X-ray Pelvis and chest, Ultra sonogram, CT / MRI and alpha feta protein.

FOLLOW UP: Monthly follow up for 1 year, then two monthly follow up till the end of the study.

METHODOLOGY:

All cases of Sacro coccygeal teratoma detected both ante natally and post natally were examined clinically.

Investigation protocol:

All the patients underwent the following investigations-

X ray chest, Ultra sonogram of abdomen, pelvis and the swelling and CECT / MRI if feasible. The tumor markers alpha feta proteins, beta HCG and LDH were done. Cardiac evaluation was done before surgery. Genetic work up was also done. The patients then went for management as per the protocol either upfront surgery or surgery after neo adjuvant chemotherapy.

The standard procedure was WIDE LOCAL EXCISION WITH COCCYGECTOMY. Type I and II were approached from the perineum. Type III and Type IV were approached initially through abdomen and followed externally as required. Neo adjuvant chemotherapy was given for inoperable tumors and metastatic tumors. Adjuvant chemotherapy with or without radiotherapy was given based on the grade of the tumor and the resection margins positivity.

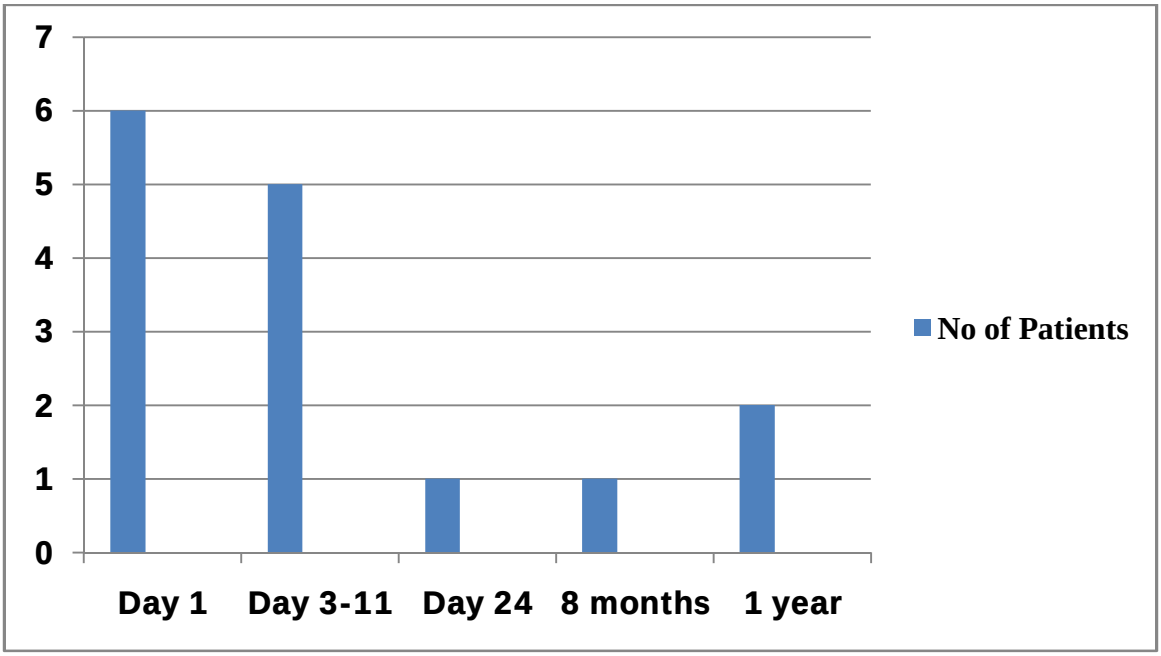
All the patients were followed till the end of 2 years of study.

OBSERVATIONS AND RESULTS:

AGE:

In our study 12 patients (80%) were diagnosed before one month of age and the remaining 3 patients were diagnosed later.

Age	No of Patients
Day 1	6
Day 3-11	5
Day 24	1
8 months	1
1 year	2

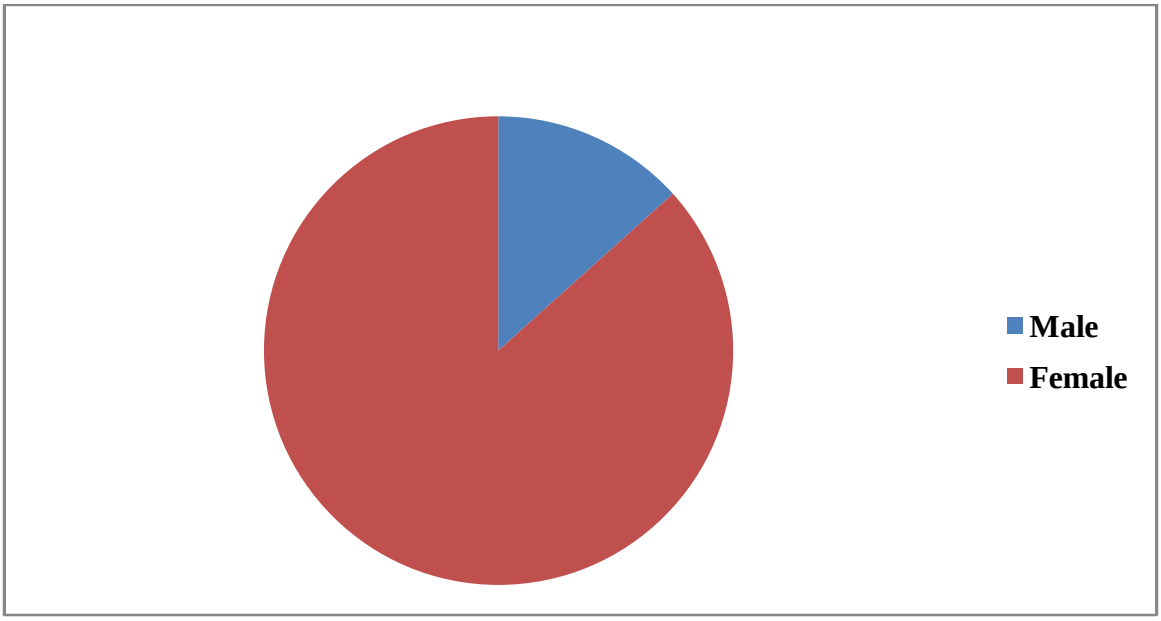


GENDER PREDILECTION:

In our study sacrococcygeal teratoma were more common in females (13) than in males (2).

The ratio M: F is 1: 6.5

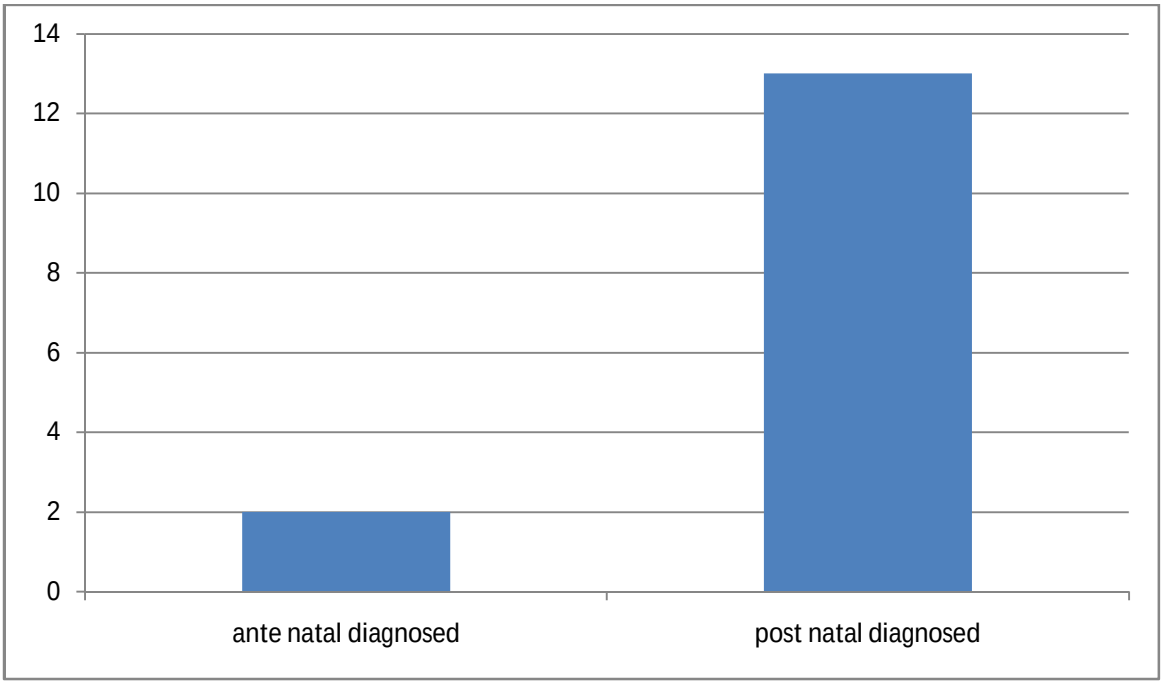
Male	2
Female	13



DIAGNOSIS:

In our study two cases were detected antenatally, and others were diagnosed postnatally. One neonate of a twin pregnancy had sacrococcygeal teratoma detected in the post natal period.

Ante natally diagnosed	2
Post Natally diagnosed	13



CLINICAL FEATURES:

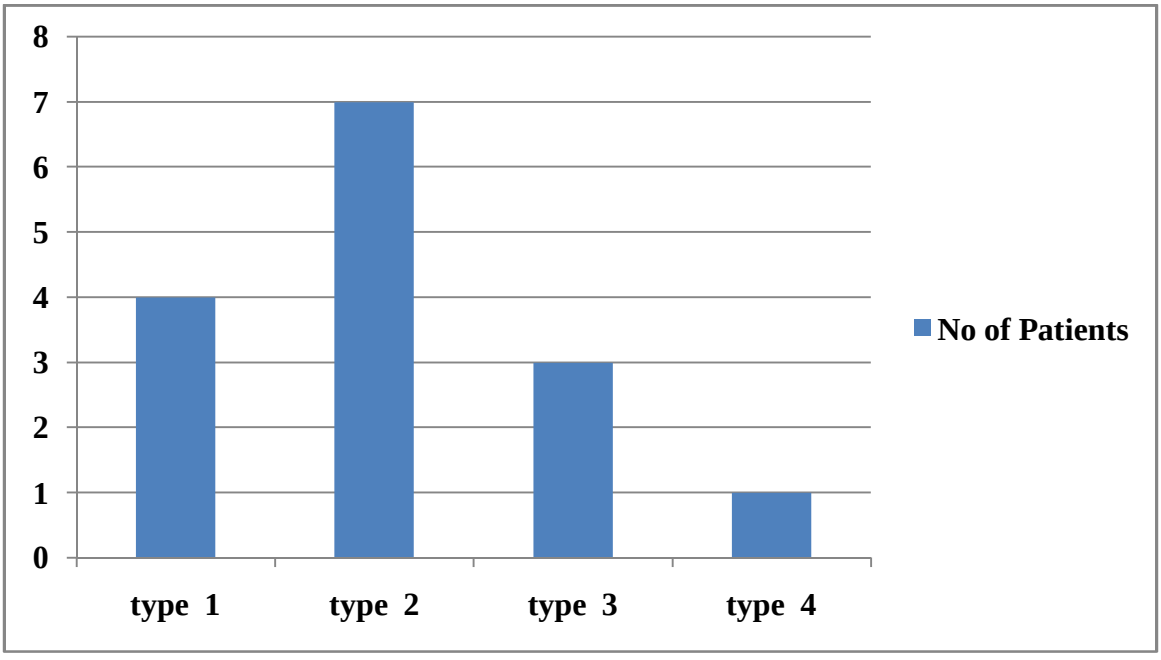
Though ante natal diagnosis is the most common mode of detection but in our study 12 patients were diagnosed in the neonatal period of which two had been diagnosed by ante natal ultra sonogram and the rest postnatally. 3 patients were diagnosed when they presented with constipation and obstructive urinary symptoms.

Diagnosis	Patients
Ante natally	2
Neonatal period	10
Infancy	3

ALTMAN TYPE:

In our study we found type 2 to be the commonest (46%) and type 4 to be the least common (6%).

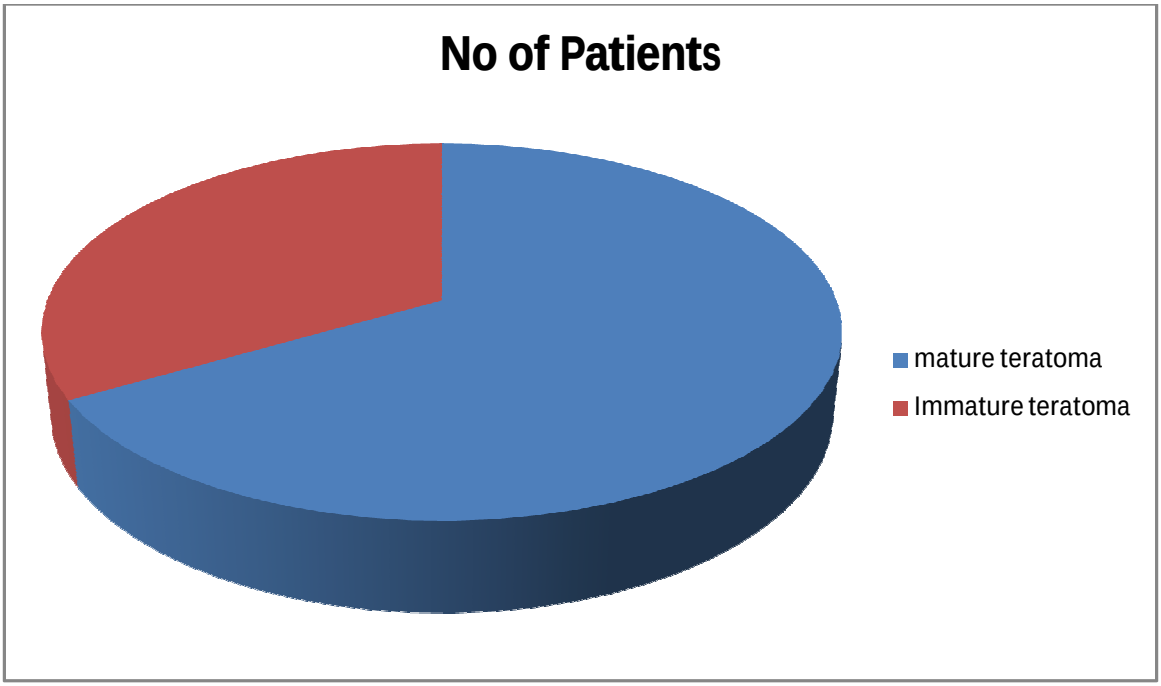
Type 1	4
Type 2	7
Type 3	3
Type 4	1



Histological types:

In our study comprised 10 patients (66 %) had mature teratoma

Mature teratoma	10
Immature teratoma	5



Management:

Primary surgery with wide local excision and coccygectomy were done in 12 patients.

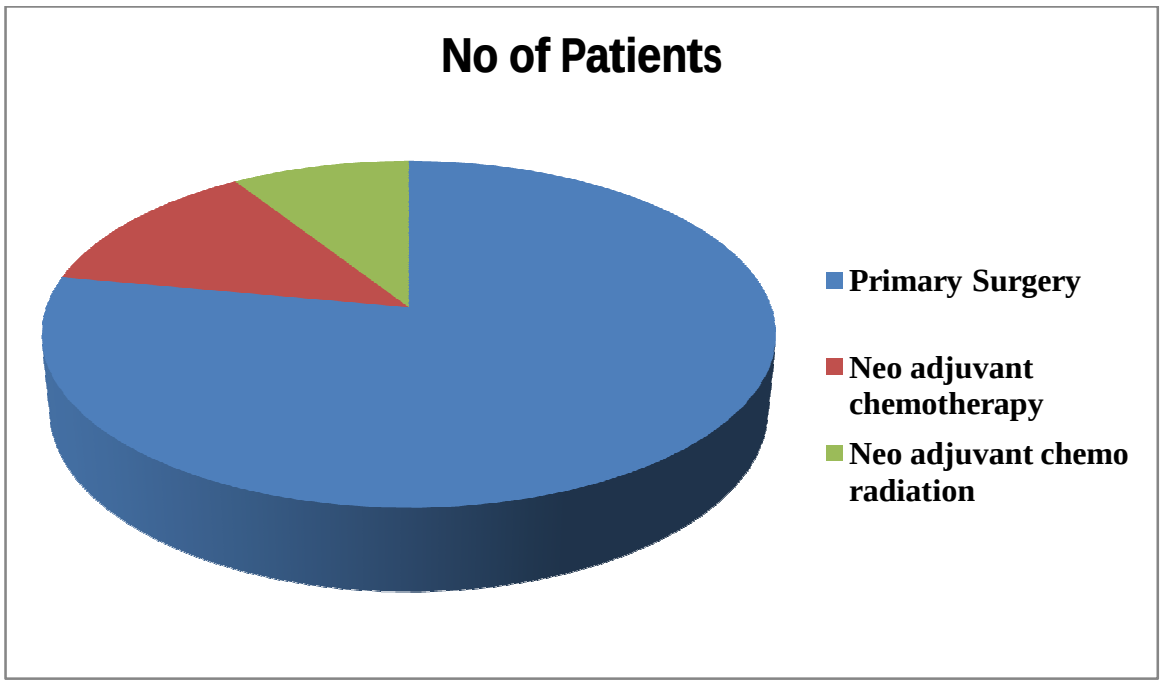
Neo adjuvant chemotherapy was started in 2 patients based on level of tumor markers and were followed with wide local excision.

Neoadjuvant chemotherapy and radiation therapy was given to 1 patient who succumbed to the illness before surgery.

The chemotherapy regime followed in our institute is PEB regime

Management methods	**No. of patients**
Primary Surgery	12
Neo adjuvant chemotherapy	2
Neo adjuvant chemo therapy and radiation therapy	1

Management Methods



Tumor markers:

Estimation of alpha feta protein, beta human chorionic gonodotropin and lactate de hydrogenase were done in all cases.

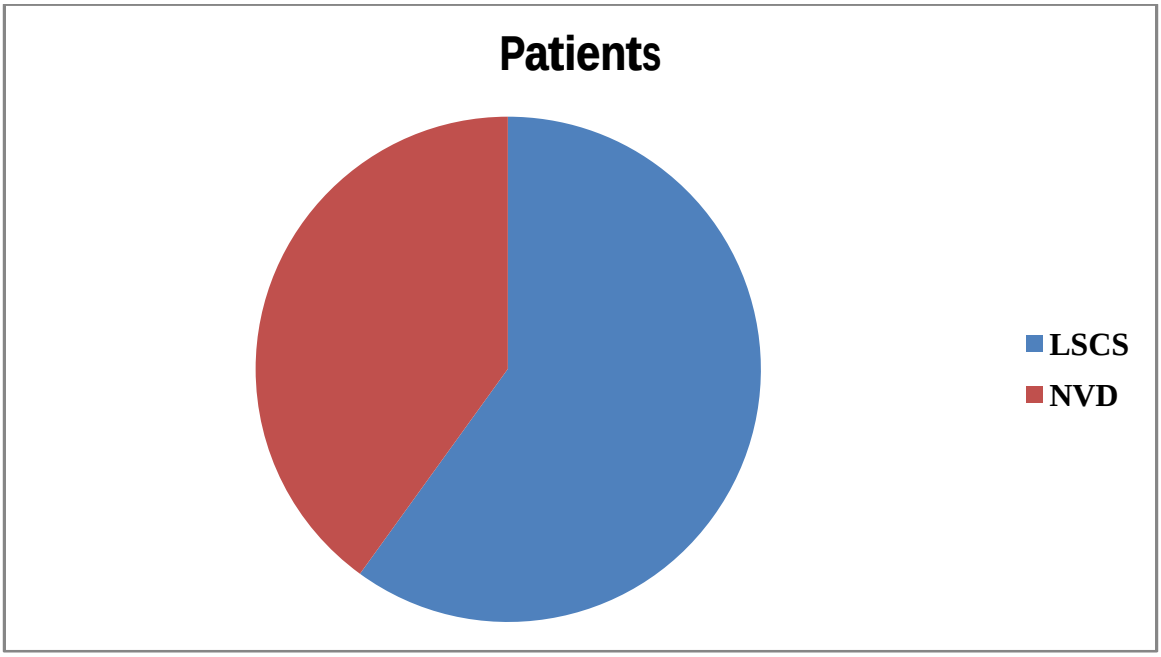
Alpha feto protein was elevated in 5 cases (33%); it was within normal range in 10 cases (66%).

Tumor markers	Increased	Normal
Alpha feta protein	5	10
Beta HCG	NIL	-
LDH	NIL	-

The presence of the tumor marker did not correlate with the tumor grade. The tumor marker was used primarily to monitor the progress of the disease and as a prognostic marker.

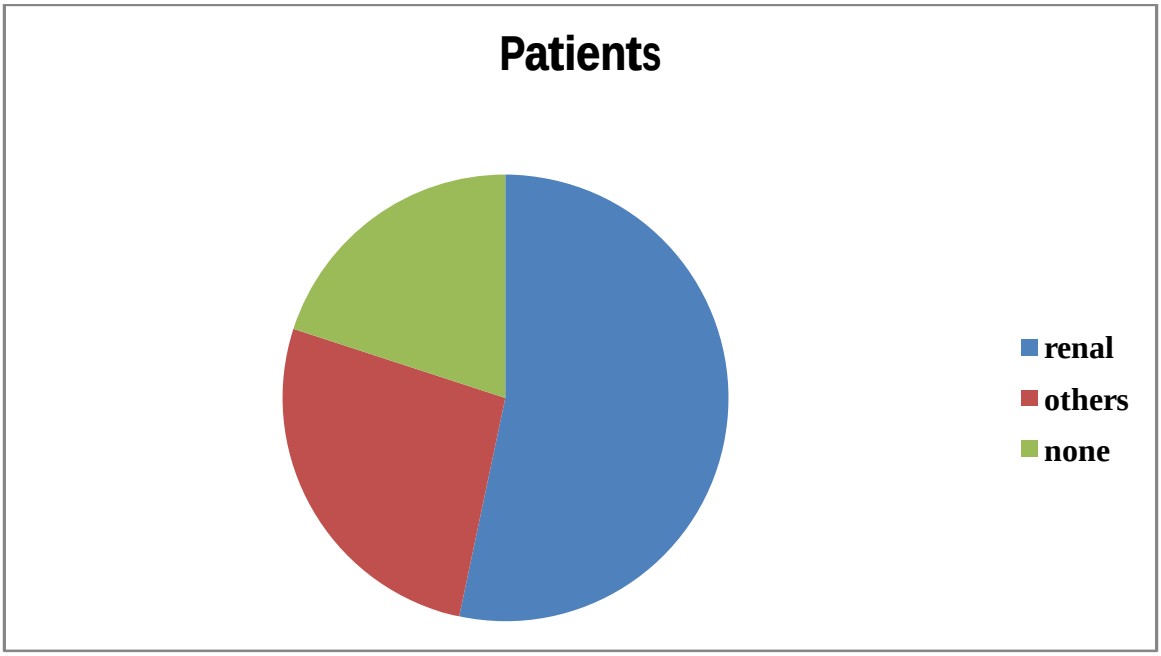
MODE OF DELIVERY:

Out of 15 patients 9 (60%) were delivered by LSCS and the rest had normal vaginal delivery.



Associated anomalies:

The most common associated anomalies were renal followed by CVS and others.



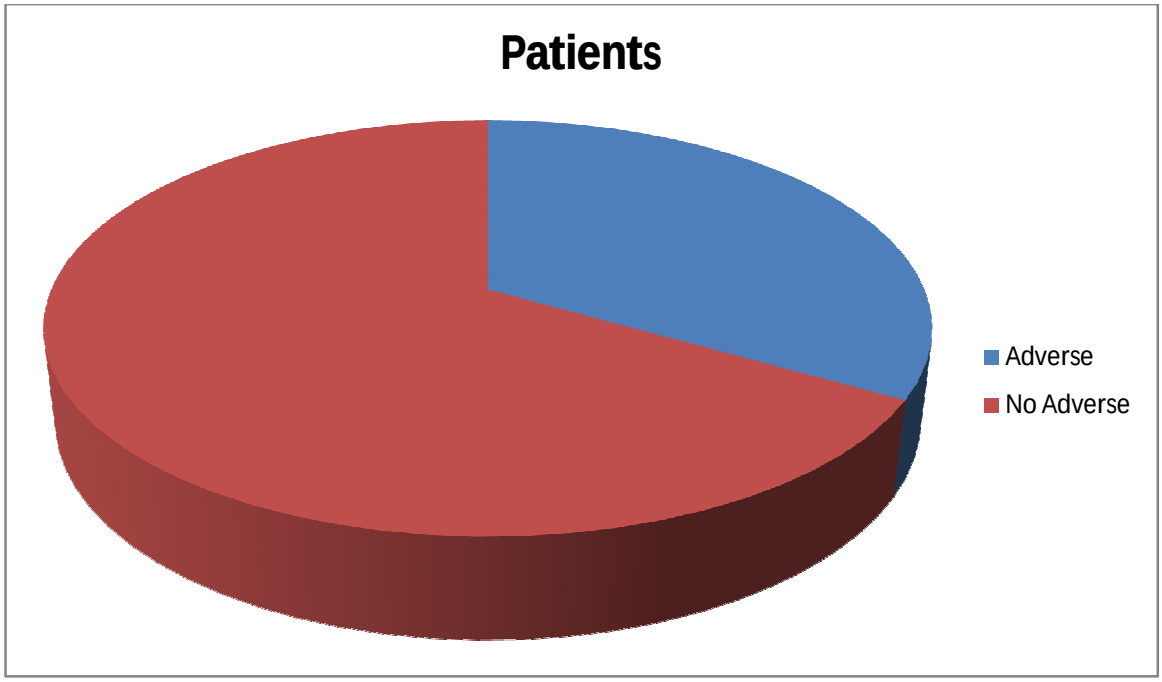
PROGNOSTIC FACTORS

The Poor prognostic factors are

1. solid tumors
2. detected early in pregnancy
3. malignant histological types
4. polyhydramnios
5. placentamegaly
6. Fetal hydrops.

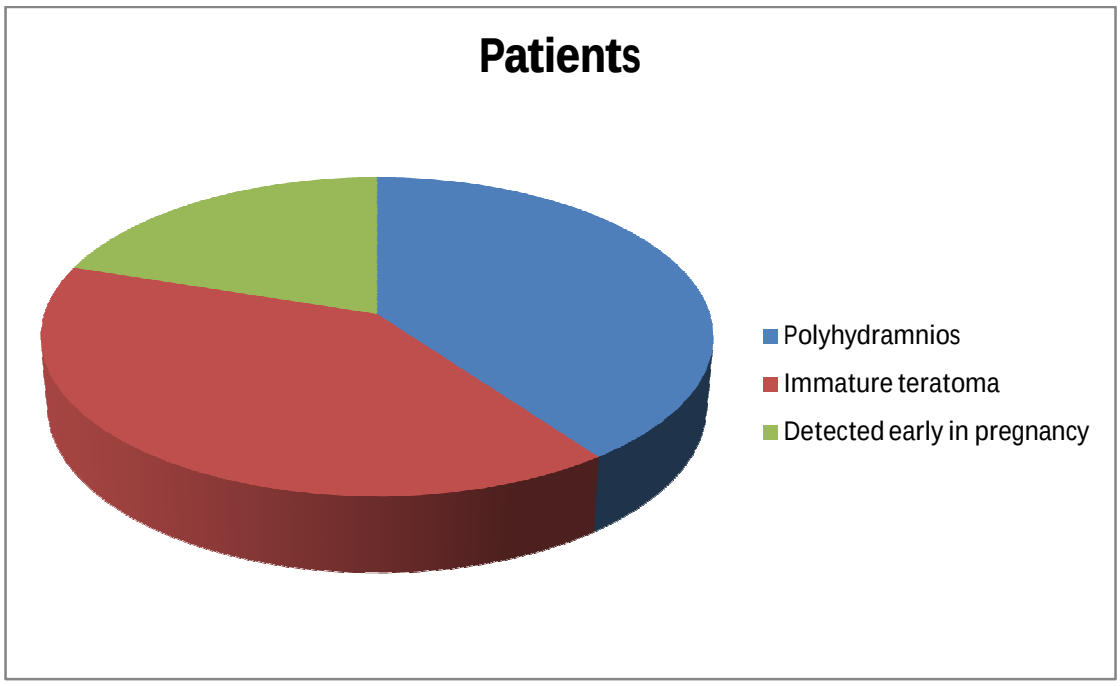
Prognosis	Patients
Poor/adverse factors	5
No adverse factors	10

PROGNOSTIC FACTORS



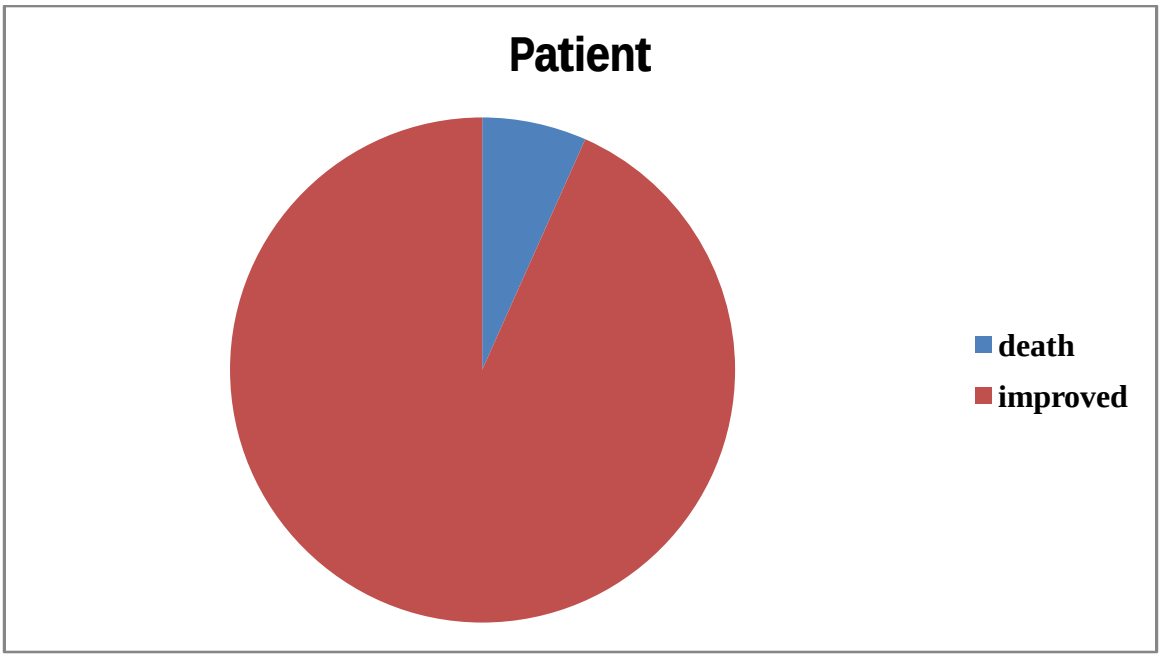
Poor prognostic factors	Patients
polyhydramnios	2
Immature teratoma	2
Detected early in pregnancy	1

Poor Prognostic Factors



MORTALITY:

During this study period one patient died during Neoadjuvant therapy while others were disease free till the completion of this study.



DISCUSSION

Discussion:

Sacrococcygeal teratoma is a congenital tumor. Incidence is 1 in 40,000 new born. It arises from the caudal end of the spine with different sizes and shapes of protrusion. Most of them are benign and well differentiated and hence have a good prognosis after complete surgical excision. [45]

It is composed of one or more germinal cell layers originating from pluripotential cells, yet it lacks complex tissular organization. The new born with Sacrococcygeal teratoma has excellent prognosis depending on the time of diagnosis and when the surgery was performed , the malignant potential of the tumor and the ease with which surgical resection was performed .[46]

Although about 75% of cases are seen in females, the exact reason for the female preponderance is not fully understood. [47,48]

Apart from age at diagnosis and treatment, and the extent of resection, the prognosis is also determined by the histological type and Altman type at the time of resection, and not the size of the tumor [49]

Follow up in patients with SCT is necessary especially during the first three years of treatment when recurrence is most likely.[50]

Extensive surgery in the pelvic and perineal region may involve disruption of nerves and muscles which supply the urinary /ano rectal sphincters which are necessary for continence. Our follow up period was for 2 years and we did not encounter any incontinence to micturition and defecation. Longer period of follow is however required, to identify late incontinence problem to micturition and defecation.

In our study females were more affected (86%) which is also in accordance with a study reported by Raney RB Jr et al from Children's hospital of Philadelphia between 1971 and 1980(66%) and also the study done by Bilik R et al Ontario, Canada. as well as by Chirdan JB et al ,Jos Nigeria who had a female preponderance of 81 %.

Their study also concluded that surgery alone was inadequate for successful management of malignant SCT which is in concordance with our study also.

Our study of 5 immature teratoma had neo adjuvant and adjuvant therapy. Our study correlates with their study for successful management of malignant SCT.

In our study Altman Type I and II were more common as also reported by Bilik R et al at the University of Toronto, Ontario, Canada from 1972 to 1990 in which they studied 36 patients.

In another study from Wakhlu A et al from Department of pediatric Surgery, King George Medical College, Lucknow, UP, India a retrospective study of 72 patients over a period of 17 years found that

1. The sex incidence was nearly equal (ours had a female preponderance)

2. There was a high proportion of Altman type IV. (We had type II)

3. 66 % presented beyond neo natal period (ours 46% presented at birth)

In a study by Perrelli L et al Catholic University of the Sacred Heart, Rome, Italy they studied 17 cases between July 1985 and December 1998 found that Benign SCT had favorable prognosis .Our study too supported this theory.

Negative prognostic factors were

1. Solid tumors,

2. Those detected early in pregnancy,

3. Malignant histotypes,

4. Polyhydramnios

5. Placentomegalay, and

6. Fetal hydrops,

Ours also corroborated with this study.

In a study by Swamy R from New Castle Medical Service observed a pre natal diagnosis in 50 % of cases were as we had only 13 %.

In a study by Chirdan JB, et al from pediatric Surgery Unit, Jos University Teaching Hospital, Jos, Nigeria between Jan 1990 and May 2008 in 38 children concluded that

1. 23 patients (60 %) presented in the neo natal period in their study were as in ours 46% presented in neo natal period.
2. Most of their patients had significant external tumors and in our study we had 46% type 2 tumors and majority of the tumors were external.
3. Their histology was mainly benign; in our study we had mature teratoma of 10 patients (66 %.) only.

In a study done by Ho KO, et al at the clinical school, University of Sidney, New South Wales, Australia between January 1996 and December 2008 had 17 infants with a diagnosis of SCT.

1. 70 % were born at term. Ours had 67 % at term child.
2. 47 % had a pre natal diagnosis. Ours had a pre natal diagnosis of 13 %.

3. Associated anomalies were seen in 41 % in their study with renal anomalies being the most common; we in ours had associated anomaly of80 % of which renal anomaly was common with 53 %.
4. Their study had an incidence of mature teratoma as 50 %, in our study we had 66% of mature teratoma.

CONCLUSION

Sacrococcygeal Teratomas are a relatively uncommon tumor affecting neonates, infants and children. Sacrococcygeal teratoma has a female preponderance.

Those presenting within a few days of birth carry a better prognosis than those detected later in infancy.

Benign Sacrococcygeal Teratomas generally have a favorable prognosis.

Benign Teratomas have a significant recurrence rate mandating follow up for more than 3 years.

Most of Sacrococcygeal teratoma present within the first few weeks of life.

Mass is the most common presenting symptom.

Early diagnosis, prompt surgical intervention, gives good results and long term survival.

Lower segment caesarian section as the mode of delivery is considered when the size of the mass is more than the Bi parietal Diameter of the fetus. When there are no other obvious indications for Lower segment caesarian section, normal vaginal delivery is considered.

Among the associated anomalies, the most common anomaly is renal anomaly.

With advancing radiological imaging, early diagnosis is possible followed by appropriate management and long term survival.

The overall survival of neonatal sacrococcygeal teratoma is high.

Tumor markers are used for monitoring the disease rather than the assessment of the grade of the disease.

Important components of management include timely diagnosis, multidisciplinary planning, (surgery, radiotherapy and chemotherapy) long term follow up, and intervention for functional sequelae.

Poor prognostic factors include solid tumor, those detected early in pregnancy, malignant histotypes, polyhydramnios, placentamegaly, and fetal hydrops.

Surgical resection alone is adequate therapy for non metastatic malignant tumor.

Cisplatin based chemotherapy has improved the survival of patients with malignant tumor.

Surgery alone is inadequate for successful management of children with malignant sacrococcygeal teratoma. Chemotherapy with PEB

(Cisplatin,Etoposide , Bleomycin) can be effective when combined with radiation, but fatal pulmonary toxicity can result from such combined therapy.

After Surgery sequelae of Sacrococcygeal teratoma tends to improve with time.

While sacrococcygeal teratoma is usually benign, recurrent malignant transformation in patient who present late and long term functional sequelae are problems that must be tackled by the care givers.

The optimal therapeutic program for children with malignant SCT is still evolving.

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Informed Consent Form

I/We,……

related to Baby/child of……

As…….. , herewith consent to enroll in the study Titled “A study of clinical profile management &outcome of Sacrococcygeal Teratoma in children” profiling conducted by Dr. K. SRIRAM as a part of his thesis for MCh, (Pediatric Surgery) post Graduation. I / We have been fully informed about the Study process, I / we have been reassured about the confidentiality of our baby’s identity. I / we know that we can withdraw from the study at any time without affecting the care and treatment of our baby/child

Witness 1

Signature:

Address:

Witness 2 Signature of parent/ Guardian

Signature: Place:

Address: Date

Sacro Coccygeal - Proforma

Personal details –

Name Age Sex

I.P.No:

Address:

D.O.A: D.O.S: D.O.D:

Antenatal scan:
Done:

Report –

Not done:

Clinical symptoms:

Ante natal Diagnosis.

Post Natal : Mass

others

Associated anamoly:

Investigation:
Ante Natal Ultrasonogram

Post Natal :

X ray No:

Chest:

Abdomen:

Spine:

Mass.

Ultrasonogram Abdomen/Cranium/mass

Echo cardiogram.

Magnetic resonance imaging (MRI)

Tumor Markers

Altman Type:

Treatment:

Surgery: Primary or adjuvant.

Chemotherapy: Adjuvant or Neo adjuvant.

Radiotherapy.

Surgical details

Intraoperative finding:

HPE report:

Adjuvant therapy.

Follow up of patient:

MASTER CHART

S.No	Name	Age	Wt	Sex	IP no	Delivery	A/N USG	MRI	Altman type	Management	HPE	Markers
1	b/o vinodhin	D10	2.8	F	750136	NVD	NIL	YES	2	Surgery	Mature	N
2	b/o jayalakshmi	D11	3.0	M	788447	LSCS	NIL	YES	1	Surgery	Mature	N
3	b/o rasigapriya	D1	3.0	F	776661	NVD	NIL	YES	2	Surgery	Mature	N
4	b/o Nalini	D1	3.2	F	767926	NVD	N	YES	1	Surgery	Mature	N
5	B/O vanilla	D6	3.0	F	762487	NVD	N	YES	2	Surgery	Mature	Increased
6	b/o Jansi rani	D1	2.0	F	751538	LSCS	YES	NIL	1	Surgery	Mature	N
7	b/o Manjula	D11	3.1	F	749890	LSCS	YES	YES	3	Surgery	Mature	N
8	b/o Rahamida	D24	3.0	F	746500	LSCS	N	YES	2	Sx & Chemo	Immature	increased
9	Divya	1 yr	7.8	F	759505	LSCS	NIL	YES	3	Neo adj Chemo & Surgery	Immature	increased
10	Rahul	1 Yr	8	M	743636	LSCS	NIL	Yes	3	Neo adj CT/RT	Immature	increased
11	b/o Padmapriyan	D3	3.0	F	767926	LSCS	N	YES	2	Surgery	Mature	N
12	B/O Rochitha	D1	3.2	F	746500	NVD	N	YES	1	Sx & CT	Immature	increased
13	b/oKajanayabraul	D1	3.0	F	739503	LSCS	N	YES	2	Surgery	Mature	N
14	b/o Kalaiselvi	D23	3.1	F	816607	LSCS	YES	YES	3	Surgery	Mature	N
15	b/o Kokila	8 M	6.0	F	788420	LSCS	NIL	YES	3	Neo Adj CT/RT	Immature	N

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